International Journal of Pediatric Otorhinolaryngology 73 (2009) 1452–1457

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Image guidance systems for minimally invasive sinus and skull base surgery in children☆,☆☆

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ARTICLE INFO

Article history:
Received 16 July 2009
Accepted 20 July 2009

Keywords:
Image-guided surgery
Paranasal sinus
Skull base
Endoscopic surgery
Minimally invasive surgery

ABSTRACT

Objective: The use of image guidance for sinonasal and skull base surgery has been well-characterized in adults but there is limited information on the use of these systems in the pediatric population, despite their widespread use. The aim of this study is to evaluate the use of image guidance systems to facilitate an endoscopic minimally invasive approach to sinonasal and skull base surgery in a pediatric population.
Methods: A retrospective cohort study was performed at a tertiary pediatric hospital. Thirty-three children presented with complications of sinusitis, tumors, traumatic, or congenital lesions of the skull base and underwent endoscopic surgery using image guidance from March 2000 to April 2007. Patient variables including diagnosis, extent of disease, and complications were extracted from paper and computer charts. Additional surgical variables including set-up time, accuracy, surgeon satisfaction index and number of uses per case were also reviewed.
Results: Twenty-eight patients (85%) underwent sinonasal surgery and five (15%) underwent skull base surgery. Indications included infectious complications of acute sinusitis ($N = 15$), neoplasms ($N = 12$), choanal atresia ($N = 4$), and cerebrospinal fluid leak ($N = 2$). Thirty-one patients (94%) required only one procedure. No surgical complications were reported. Surgeon satisfaction, mean accuracy and number of uses per procedure increased over time ($p < 0.05$).
Conclusions: Image guidance systems are safe and effective tools that facilitate a minimally invasive approach to sinonasal and skull base surgery in children. Consistent with adult literature, usage and surgeon comfort increased with experience. The additional anatomical information obtained by image guidance systems facilitates a minimally invasive endoscopic approach for sinonasal and skull base pathologies.

1. Introduction

Skull base and sinus surgery in children poses particular risks and challenges. Throughout development, the nasal and paranasal spaces continue to grow and change dramatically. In addition, there are a vast array of pathologies that can occur in this area, including congenital abnormalities, benign and malignant neoplasms, infections and traumatic injuries. Image-guided surgery (IGS) has the potential to offer substantial benefit for procedures in this area, specifically in the treatment of complications of acute sinusitis and for evaluation and treatment of tumors, traumatic and congenital abnormalities. In the adult literature, IGS has been shown to decrease the incidence of major complications and allow a more complete and effective surgery [1]. The American Academy of Otolaryngology Head and Neck Surgery formulated a consensus of expert opinion on indications for image-guided surgery [2] that includes revision surgery, distorted anatomy, skull base defects, and sinonasal neoplasms. IGS is rapidly becoming the standard of care for adult revision sinus surgery. Despite the intense research on image-guided surgery in adults, there is very little published literature on the use of this technology in the pediatric population. In children with smaller and more variable anatomy, the risks and benefits of IGS should be studied further. The goal of this study is to report our experience using IGS in the management of complicated

☆ This paper was presented in poster format at the Eastern Section Meeting of the Triological Society, Boston, MA 02114, USA, January 23–25, 2009.
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doi:10.1016/j.ijporl.2009.07.017

Table 1
Distribution of diagnoses in the series.

Etiology	Diagnoses	Total number
Infectious	Complications of acute sinusitis	15
Neoplastic		12
Benign	Anterochoanal polyp	1
	Juvenile nasopharyngeal angiofibroma	3
	Neurofibroma	1
	Glioma	3
	Teratoma	1
	Ossifying fibroma	1
	Langerhan's cell histiocytosis	1
Malignant	Rhabdomyosarcoma	1
Congenital	Choanal atresia, CSF leak	5
Traumatic	CSF leak	1
Total		33

sinus and skull base pathologies with regard to indications, outcomes, complications, and surgeon comfort.

2. Methods

Thirty-three children underwent endoscopic sinonasal or skull base surgery for complications of sinusitis, neoplasms, or traumatic or congenital abnormalities of the skull base using IGS between March of 2000 and April of 2007. Information about patient characteristics including age, gender, diagnosis, outcome, complications, and need for additional surgery was extracted from patient charts. Patients undergoing routine endoscopic sinus surgery for chronic sinusitis using IGS were specifically excluded from this study. The LandmarX®system (Xomed® Medtronic®) was used in all procedures. Seven sensors were placed on the patient's forehead according to the LandmarX protocol. Reference points were then selected and calibrated. The setup time, accuracy, number of times used during each surgery and the ''How Helpful Index'' (HHI) were recorded. Correlations over time were obtained using regression analysis and a t-statistic and p-value were calculated. A probability less than 0.05 was considered significant.

3. Results

The diagnostic entities are summarized in Table 1, including complications of acute sinusitis, benign and malignant neoplasms, choanal atresia and cerebrospinal fluid (CSF) leak. Five patients underwent skull base surgery and 28 underwent sinonasal surgery (including one patient with an orbital mass) using image guidance. Patient characteristics are listed in Tables 2A and 2B. There were 20 males (61%). The mean age at time of surgery was 10.3 years (range 9 days to 25 years, median 11 years).

Table 2A
Patient characteristics: complications of acute sinusitis.

Patient	Age and gender	Diagnosis	Surgery
1	6yF	Subperiosteal abscess	Unilat MA, AE
2	7yM	Subperiosteal abscess	Unilat MA, AE
3	7yM	Pre-septal cellulitis, recurrent dacrocystorhinitis	Bilat MA, AE, DCR
4	7yM	Orbital cellulitis, subperiosteal abscess	Unilat MA, TE, orbital decompression
5	9yF	Subperiosteal abscess	Bilat MA, TE, frontal sinusotomy
6	9yM	Subdural abscess	Bilat MA, TE, frontal sinusotomy
7	10yM	Subdural abscess, Pott's puffy sinusitis	Unilat MA, TE, frontal sinusotomy, FST
8	11yF	Pre-septal cellulitis	Unilat MA, TE
9	12yM	Subperiosteal abscess	Unilat MA, AE
10	12yM	Orbital cellulitis, subperiosteal abscess	Bilat MA, TE, frontal sinusotomy
11	14yM	Pott's puffy sinusitis	Bilat MA, TE, FST
12	14yM	Frontal sinusitis	Endoscopic frontal sinusotomy, FST
13	14yF	Frontal sinusitis with dehiscence	Unilat MA, AE, frontal sinusotomy
14	17yF	Meningitis	Bilat MA, TE, S
15	18yM	Cavernous sinus thrombosis	Bilat MA, TE, S

AE – anterior ethmoidectomy, DCR – dacrocystorhinostomy, FST – frontal sinus trephine, MA – maxillary antrostomy, S – sphenoidotomy, TE – total ethmoidectomy.

Table 2B
Patient characteristics: neoplasms, inflammatory, congenital and traumatic lesions.

Patient	Age and gender	Diagnosis	Surgery
1	16yM	Juvenile nasal angiofibroma	AE, S, excision of mass
2	17yM	Juvenile nasal angiofibroma	Unilat MA, TE, excision of mass
3	18yM	Juvenile nasal angiofibroma	Excision of mass
4	0.1yF	Nasal glioma	Excision of mass
5	1yM	Nasal glioma	Excision of mass
6	1.8yM	Teratoma of masticator space	Transoral excision of mass
7	4yF	Infratemporal fossa desmoid	Biopsy of infratemporal fossa mass
8	5yM	Histiocytosis of orbit	Excision of orbital mass
9	7yM	Anterochoanal polyp	Unilat MA, AE, Excision of mass
10	10yF	Rhabdomyosarcoma infratemporal	Excision of infratemporal fossa mass
11	13yF	Ossifying fibroma	External frontoethmoidectomy, midface osteotomy
12	25yF	Neurofibromatosis Type 1	Biopsy of infratemporal skull base mass
13	1yM	Congenital CSF leak	CSF leak repair
14	13yM	Traumatic CSF leak	CSF leak repair
15	1.5yM	Choanal atresia	Endoscopic choanal atresia repair
16	6yF	Choanal atresia	Endoscopic choanal atresia repair
17	15yF	Choanal atresia	Endoscopic choanal atresia repair
18	9yF	Choanal atresia	Endoscopic choanal atresia repair

AE – anterior ethmoidectomy, CSF – cerebrospinal fluid, MA – maxillary antrostomy, S – sphenoidotomy, TE – total ethmoidectomy.

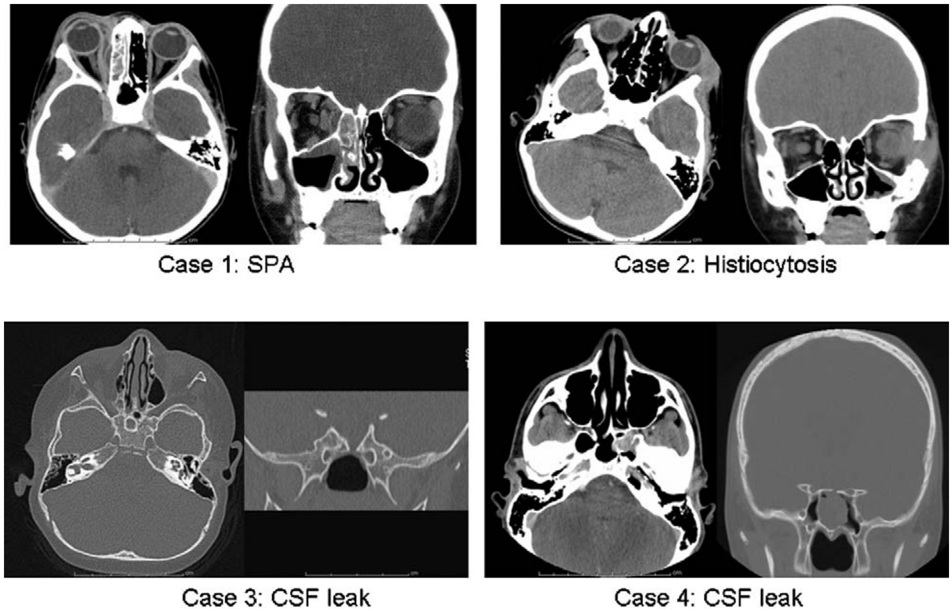

Fig. 1. Selected cases with infectious, neoplastic, congenital and traumatic etiologies. Computed tomography images are shown in the axial and coronal planes for four cases treated with image-guided endoscopic sinus and skull base surgery. Case 1 shows a subperiosteal abscess (SPA) adjacent to opacified left ethmoid air cells. Case 2 depicts Langerhans cell histiocytosis of the left orbit. Cerebrospinal fluid leaks requiring surgical repair of the skull base are shown in Case 3 (congenital) and Case 4 (traumatic).

Fifteen patients underwent endoscopic sinus surgery for complications of acute sinusitis (Table 2A). Indications for surgery included subperiosteal abscess, orbital cellulitis and abscess, dacrocystorhinitis, cavernous sinus thrombosis, meningitis, and subdural abscess. One patient with a frontal subperiosteal abscess (Pott's puffy sinusitis) and subdural abscess initially underwent endoscopic sinus surgery followed by frontal sinus trephination with nasal endoscopy two weeks later for persistent disease. Eleven patients were diagnosed with benign tumors of the head and neck (Table 2B) and one patient with a malignant tumor, a rhabdomyosarcoma involving the infratemporal fossa.

Fig. 1 shows axial and coronal computed tomography (CT) images for four patients with varied pathologies. Case 1 is a patient with opacification of the right ethmoid air cells with disease extending through the lamina papyracea to form a subperiosteal abscess in the orbital space adjacent to the medial rectus muscle.

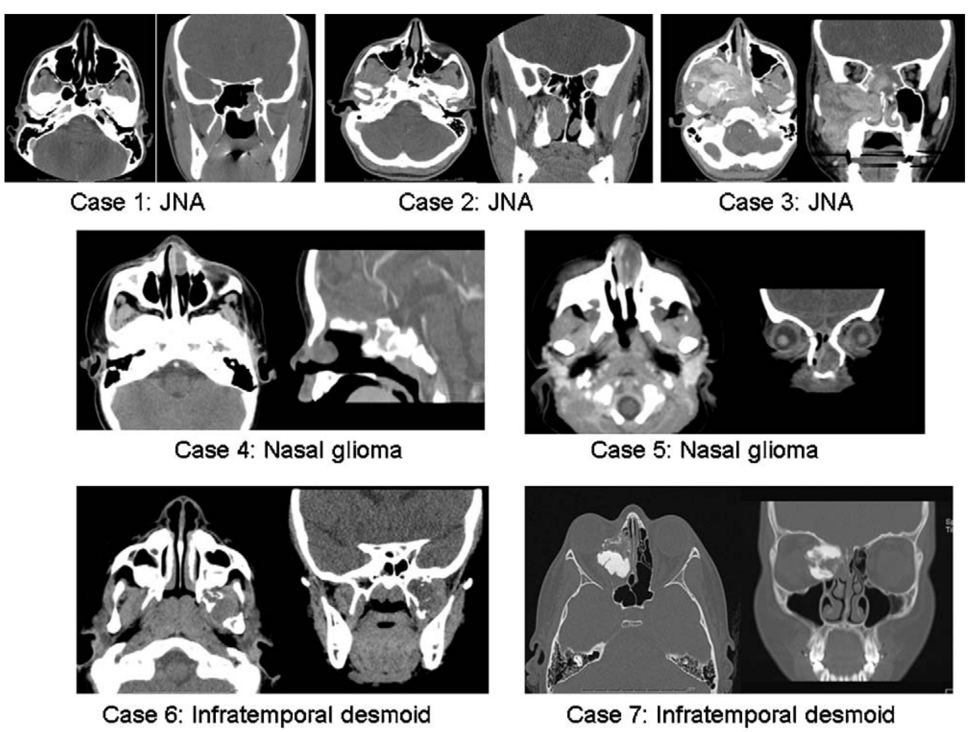

Fig. 2. Neoplastic etiologies. Cases 1–3 depict juvenile nasal angiofibromas. Cases 1 and 2 are confined to the nasal cavity and paranasal sinuses, while Case 3 shows a JNA extending laterally to involve the skull base and soft tissues of the face. Cases 4 and 5 show anterior nasal gliomas that were successfully removed using image-guided endonasal surgery. Cases 6 and 7 show infratemporal desmoid tumors, encroaching upon the orbit in Case 7.

Table 3
"How Helpful Index" (HHI) description of terms.

HHI score	Significance
1	Not helpful
2	Slightly helpful
3	Helpful
4	Very helpful

Table 4
Evolution of feasibility and reliability parameters over time.

Year	Mean setup time (min) (p = .59)	Mean accuracy (mm) (p = .02)	Mean HHI (p = .02)
2000	6.67	2	3.3
2001	9	2	3.2
2002	10	2.2	3.125
2003	6	1.9	3.3
2004	5	1.9	4
2005	10	1	4
2006	10	1	4

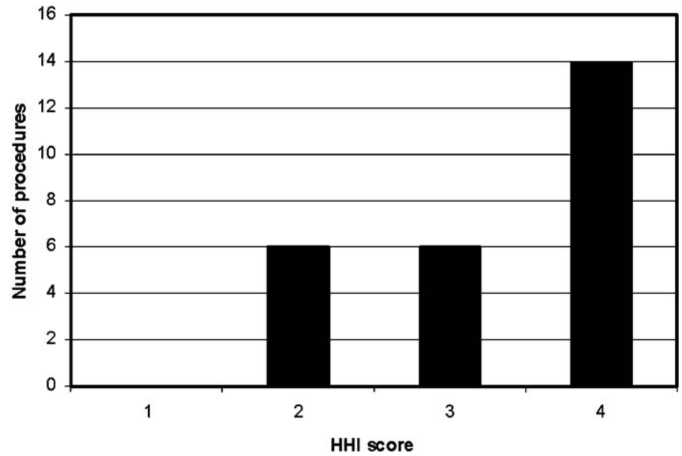

Fig. 4. Number of procedures ranked at each HHI score. No case was rated a 1, "not helpful". The majority of cases were scored as a 4, "very helpful", the highest rating possible.

Case 2 is a left orbital histiocytosis that was successfully resected using image guidance. Two patients underwent endoscopic CSF leak repair using IGS (Cases 3 and 4). Case 3 illustrates a congenital defect in the skull base resulting in symptomatic CSF leak. Case 4 shows a defect in the roof of the sphenoid sinus after traumatic head injury in a 13 year-old male. Both cases were repaired successfully with no evidence of CSF leak on follow-up. Four patients underwent endoscopic choanal atresia repair (not shown).

Fig. 2 depicts seven examples of neoplastic disease treated with a minimally invasive approach using image guidance systems. Cases 1–3 involve young male patients with a diagnosis of juvenile nasal angiofibroma. Cases 4 and 5 illustrate nasal glioma neoplasms treated with full excision endoscopically using image guidance. Cases 6 and 7 are infratemporal desmoid tumors that were biopsied safely with the help of an image guidance system.

Two patients required repeat surgery. One patient with a JNA underwent a second procedure 1 year later for recurrence. One patient required an open lynch procedure for persistent subperiosteal abscess. Two patients underwent surgery at other institutions prior to being seen in our department. Thirty-one out of 33 patients (94%) were free of disease after one procedure at our institution. The total number of surgical procedures was 48. There were no minor or major complications in the perioperative period. Three patients were lost to follow-up. For the remaining 27 patients, the mean follow-up time was 2 years (range 9 months to 3.5 years).

Image guidance systems were used for all 48 procedures and rated by the operating surgeon in 26. Information was gathered regarding setup time, accuracy, number of times used per procedure, and "How Helpful Index" (HHI, defined in Table 3). Data averaged per year is depicted in Table 4. Pooling all data together, the mean setup time was 8.6 min (range 5–20 min) and the mean accuracy was 1.97 mm (median 2 mm, range 1–5 mm). Setup time did not vary significantly from the beginning to the end of the series ($p > .05$); however the improvement in mean accuracy over time was statistically significant ($p = .02$). The HHI (mean 3.33, range 2–4) also increased over the course of the study ($p = .02$). Image guidance was used less than 5 times per operation in 4 cases, from 5 to 10 times in 3 cases, from 10 to 15 times in 1 case and 15 or more times in 16 cases. Over the course of the study the number of uses per procedure increased as shown in Fig. 3. In the majority of cases the HHI was scored in the highest possible ranking, "very helpful" (Fig. 4). There was no significant difference in HHI according to diagnosis (Fig. 5), since all diagnostic categories scored IGS highly. No correlation was found between surgeon and setup time, accuracy, and HHI ($p > 0.05$).

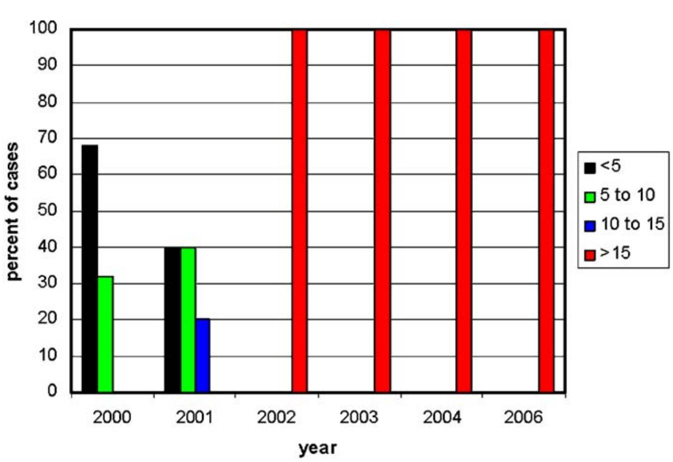

Fig. 3. Number of times the IGS system was used per procedure according to year. Starting in 2002, the IGS system was used more than 15 times per procedure ($p < 0.01$) in 100% of cases.

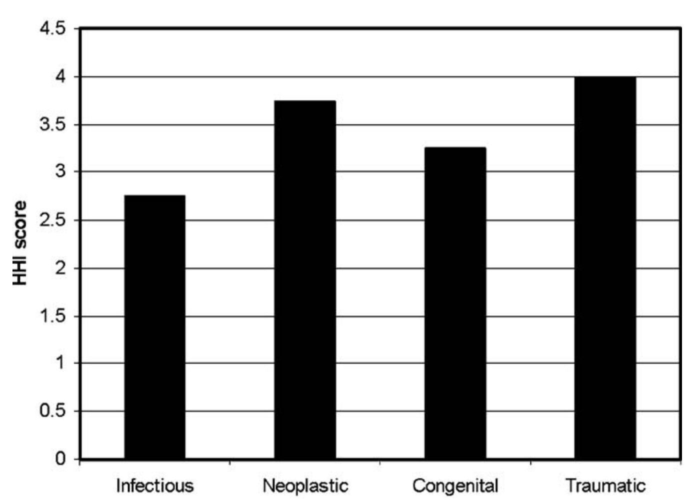

Fig. 5. HHI average in each category of disease. There was no statistically significant difference in HHI score across disease entities.

4. Discussion

The first image guidance surgical system for use in sinus surgery was developed in Germany in 1986 [3]. Rhinologists were the first to express meaningful interest in IGS [4,5]. Although a great deal of research has been generated over the past 20 years on image guidance in adult otolaryngology, there is very little data to direct clinical practice regarding the use of this technology in pediatric sinonasal and skull base surgery. The benefits attributed to image-guided surgery include assistance in defining landmarks, avoidance of complications, increasing surgeon confidence resulting in a more thorough procedure, and facilitating a minimally invasive approach with decreased need for facial or intraoral incisions. In 2002, the American Academy of Otolaryngology-Head and Neck Surgery published a consensus statement with indications for using image guidance in adult sinus surgery [2]. In particular, IGS is encouraged for revision surgery, cases when the anatomy may be distorted, for disease abutting the skull base or orbit, and for neoplastic conditions. In children, IGS can provide a similar benefit for a variety of challenging surgical cases. The anatomy of the paranasal sinuses and skull base changes drastically over the course of development. The surgical field is considerably smaller than in the adult and requires a high level of caution during surgical intervention [6,7]. In addition, congenital anomalies and acute inflammation can further distort anatomy and obscure surgical landmarks.

In the present study, routine sinus procedures were excluded in order to emphasize the use of image guidance for complications of acute sinusitis, neoplastic, congenital and traumatic etiologies. In each instance, the nasal anatomy and typical surgical landmarks may be elusive and atypical. Achieving a minimally invasive approach in children minimizes unfavorable facial scarring and does not interfere in facial growth and development [8]. Published reports favor the use of image guidance systems as equally or more beneficial than open approaches in pediatric sinonasal and skull base surgery for chronic sinusitis and neoplasms. In addition, select case reports highlight novel uses for congenital and developmental disorders in children [7,9,10]. Despite the logical benefits of using image guidance in this population, the safety and efficacy of image-guided surgery in pediatric otolaryngology has not been well documented. Clearly further research on this topic would be beneficial to the pediatric otolaryngology community. However, due to the low complication rate in endoscopic sinus surgery, a prohibitively large patient series would be required to demonstrate any direct benefit to patients [11–13]. Currently in children as well as in adults, prospective randomized controlled trials designed to evaluate the complications of IGS are neither feasible nor ethical [1,14].

Several published retrospective reports have compared endoscopic to open approaches and have found favorable results [15,16]. For example, Pryor et al. [15] reported a series of patients treated for JNA with either an open or endoscopic approach and found that those treated endoscopically had a shorter length of stay, lower risk of complications, and lower rate of recurrence. Rahbar and colleagues [16] found that in the treatment of orbital subperiosteal abscess in children, an external approach was associated with an increase in length of stay compared to endoscopic surgery. Thus although prospective randomized control studies are not available, retrospective data supports the use of IGS for the indications presented here. Without additional data on patient safety, it is important to emphasize that IGS does not replace the operating surgeon's experience and should be used as an adjunct in facilitating clinical intraoperative decision making [11,17].

With any new surgical device or technique, both reliability and feasibility must be evaluated. In this case series, reliability was addressed by the rate of surgical complications, the accuracy of the equipment, and the need for additional or open operation. The accuracy improved by a small but significant amount over time. No surgical complications occurred in this series; however, as mentioned above, limited conclusions can be drawn regarding complication rate without an adequate comparison group. The vast majority of published reports on endoscopic sinus surgery in children cite a very low complication rate [9,15,18–20] as in the present case series. The use of IGS may allow for a more complete procedure due to definition of landmarks, particularly for acute or chronic infectious processes. However, it did not prevent multiple surgical procedures in pathologies that are prone to recurrence, such as JNA. In revision surgery, IGS becomes even more important and beneficial.

The criteria used to evaluate feasibility of image-guided surgery included setup time, number of times IGS was used per procedure, and the HHI. The mean setup time was less than 10 min each year and was not correlated with the amount of time the surgeon had been using this equipment (Table 3). This amount of time for setup and testing of the device was not prohibitive or excessive and did not negatively impact the surgeon's decision as to whether to use the technology or not. The number of times the equipment was used per case and the HHI increased over time. This is possibly related to the learning curve involved in becoming comfortable with the technology. IGS has been shown to increase surgeon confidence [17] and the time evolution noted in our study suggests that the more IGS is used, the more helpful it can be. After an initial increase over the first 2 years, usage and HHI both reached a plateau at the highest rating on each respective scale. A similar increase and plateau was reported by Metson et al. [17] that was interpreted as an initial enthusiasm for the device. The plateau in our data at the highest possible rating suggests that further increases may have been detected with an extended rating system. Alternatively, this trend could be due to initial scheduling of cases at all levels of difficulty, followed by a strategy of selecting only the more challenging cases once the system is well known. Later in the time course of the present study most patients underwent surgery for complicated tumors, CSF leak repair and choanal atresia repairs.

The present study can be compared to prior reports on the use of IGS in children. In 2002, Postec et al. [9] reported on the use of a computer-assisted navigation system in the management of pediatric sinonasal surgery. The mean setup time and accuracy is similar to that found in the present study. They additionally found that indications for use increased as surgeons became more comfortable with the device. Our current study expands on this preliminary report by broadening the surgical indications to include complications of acute sinusitis and skull base pathologies such as CSF leak. In addition, the present study includes a more specific analysis of surgical factors such as the HHI and number of times IGS was used per procedure. Another recently published study [21] similarly found that IGS was safe and effective for use in the pediatric population for chronic sinusitis and polyposis in addition to a variety of other pathologies. The present study specifically excluded those patients undergoing sinus surgery for chronic sinusitis in order to focus on more acute complex cases. As such, the indications for the use of IGS in pediatric sinonasal and skull base pathologies are further expanded.

The present study is limited by the small number of patients and the retrospective nature of the review. Despite these limitations there are contributions to the study of image-guided surgery in children, including the types of pathologies that can be treated, outcomes, complications, and surgeon related information including trends in usage over time.

Image guidance systems are safe and effective tools that facilitate a minimally invasive approach to sinonasal and skull base surgery in children. Recommendations for the use of IGS for

pediatric sinonasal and skull base procedures can and should parallel indications in adults; namely revision cases, tumors and traumatic injuries. In addition we would recommend the use of IGS for acute infectious complications and congenital lesions, in which the anatomy and landmarks can be expectedly distorted.

Funding

Interdepartmental.

Conflict of interest statement

No author has any financial or personal conflicts of interest to report.

Acknowledgments

The authors would like to thank Ms. Kristen Lowry and Ms. Amanda Coburn for their assistance with data collection and preparation of the manuscript.

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